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Case Report
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Reduction in Vision as the Initial Presentation of Metastasized Primary Adenoid Cystic Carcinoma of the Lung: A Case Report

Alharbi A*, Alramyan A, Alsaleh H, Paramasivam M and Alotaibi B

Department of Medicine, Division of Pulmonology, King Saud University Medical City, King Saud University, Riyadh, KSA

Introduction

Adenoid cystic carcinoma is primarily a malignant tumor originating in the salivary glands [1]. Primary adenoid cystic carcinoma of the lung is rare, accounting for only 0.09% to 0.2% of all lung cancers [2]. Most cases of pulmonary adenoid cystic carcinomas occur in the trachea and major bronchi and majority present with local symptoms [3]. In this report, we present a rare case of adenoid cystic carcinoma of the lung presenting with reduction in vision in a young female patient.

Case Presentation

A 31-year-old female, not known to have any medical disorders, presented to an ophthalmology center with progressive blurring of vision of left eye over a period of one year. No photosensitivity, redness, diplopia, or any discharge. No neurological deficits. Denied chest pain, cough or sputum, fever, weight loss or loss of appetite. She is a passive smoker. No relevant family history.

Retinal examination showed abnormal Fundus; inferior retinal detachment with choroidal mass, see Figure 1.

Physical examinations revealed no remarkable findings. Laboratory work up was essentially unremarkable.

Initial chest X-ray showed mass-like opacity behind the heart (Figure 2). Subsequently contrast enhanced Computer Tomography (CT) scan of the thorax was performed which showed left lower lobe irregular shaped mass encasing and partially attenuating the left lower lobe bronchus, the appearance is concerning for a neoplastic process (Figure 3).

CT guided biopsy of the lesion was then performed, and careful histopathological examination suggests that the underlying malignant lesion is lung Adenoid cystic carcinoma (Figure 4). In Immunohistochemistry analysis the tumor cells exhibited a positive expression of c-kit (multifocal), CK7, CK5/6 with no expression of S100, P63, TTF-1, Napsin A, SMA, Synaptophysin, Chromogranin, Calp, HMBG-45 and Melan A. Proliferation index Ki67 is high.

To assess the extent of the disease, PET scan, bone scan and MRI brain was performed (Figures 5-7).

Patient was referred to medical oncology service for further treatment. Patient had Eastern

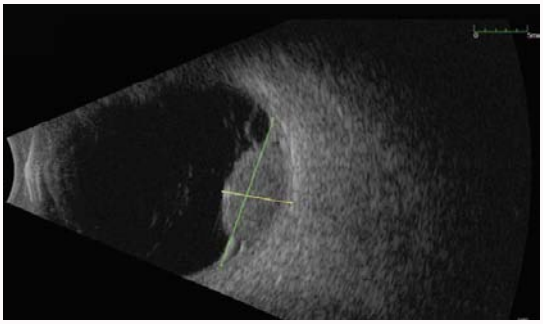

Figure 1: B-SCAN, A well circumscribed dome shaped lesion at macula and inferior to macular area with shallow focal retinal detachment at base of lesion.

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Correspondence:
Abdullah Alharbi, Department of Medicine, Division of Pulmonology, King Saud University Medical City, King Saud University, Riyadh, KSA, E-mail: dr_alharbi99@hotmail.com

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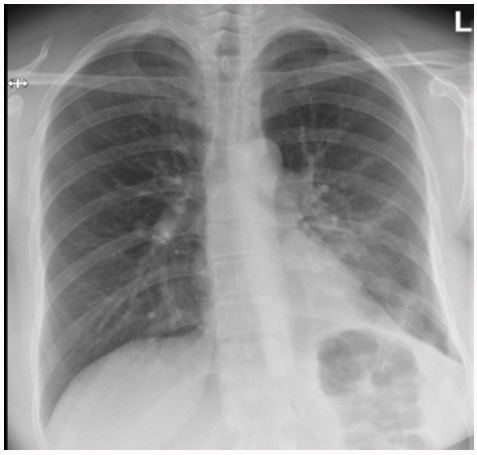

Figure 2: Initial CXR showed mass like round opacity at the left lower zone behind the heart, with elevated left hemidiaphragm.

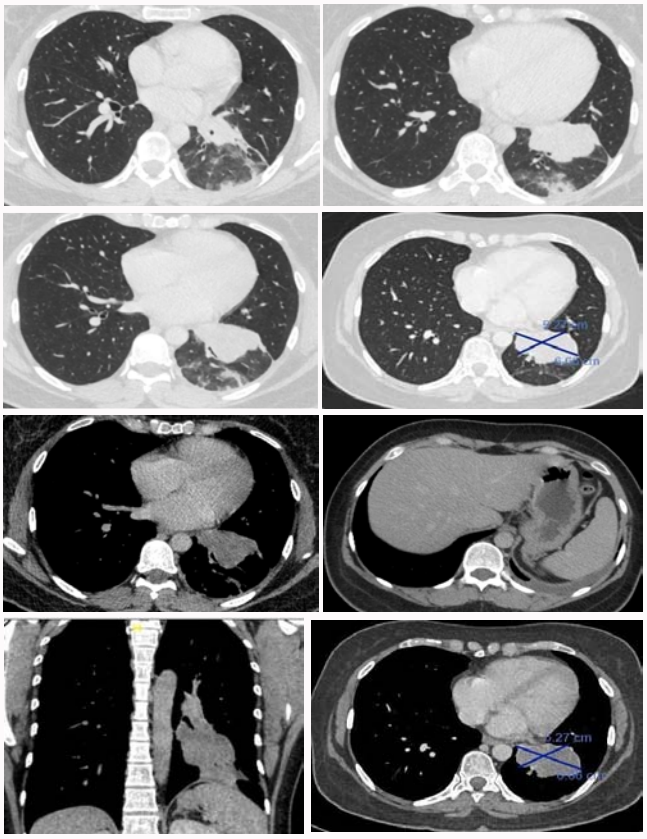

Figure 3: CT chest shows Left lower lobe irregular shaped mass measuring 6.6 cm × 5.2 cm encasing and partially attenuating the left lower lobe bronchus.

Cooperative Oncology Group (ECOG) class 1, started then to complain from pain in the back and hips. Her case was discussed in the tumor board and plan was to start cisplatin plus gemcitabine-based chemotherapy and to manage her back and hip pain with palliative radiotherapy. Pembrolizumab was unavailable at the time of starting chemotherapy.

Repeat PET-CT, CT chest, abdomen and pelvis was performed after receiving 4 cycles of chemotherapy and approximately after 3 months from the initial PET scan. Unfortunately, repeat PET-CT

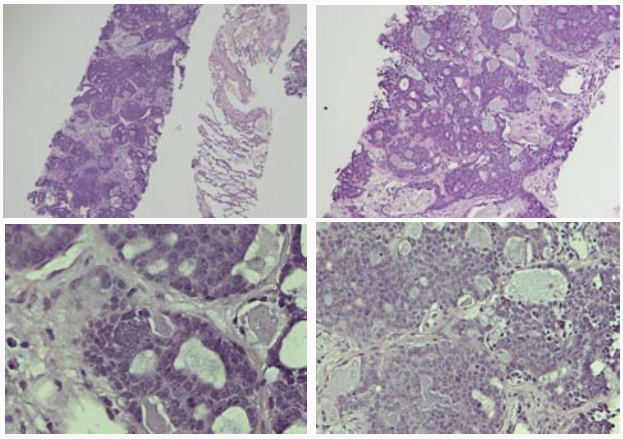

Figure 4: Microscopic examination showed an invasive malignant proliferation with a mainly cribriform architecture associated with small clusters, tubes and solid proliferation. The tumor cells were medium to large in size with abundant eosinophilic cytoplasm, enlarged atypical nucleus with prominent nucleoli. Mitotic figures are obvious. The proliferation is imbedded in a mucoid matrix, highlighted by mucicarmine stain.

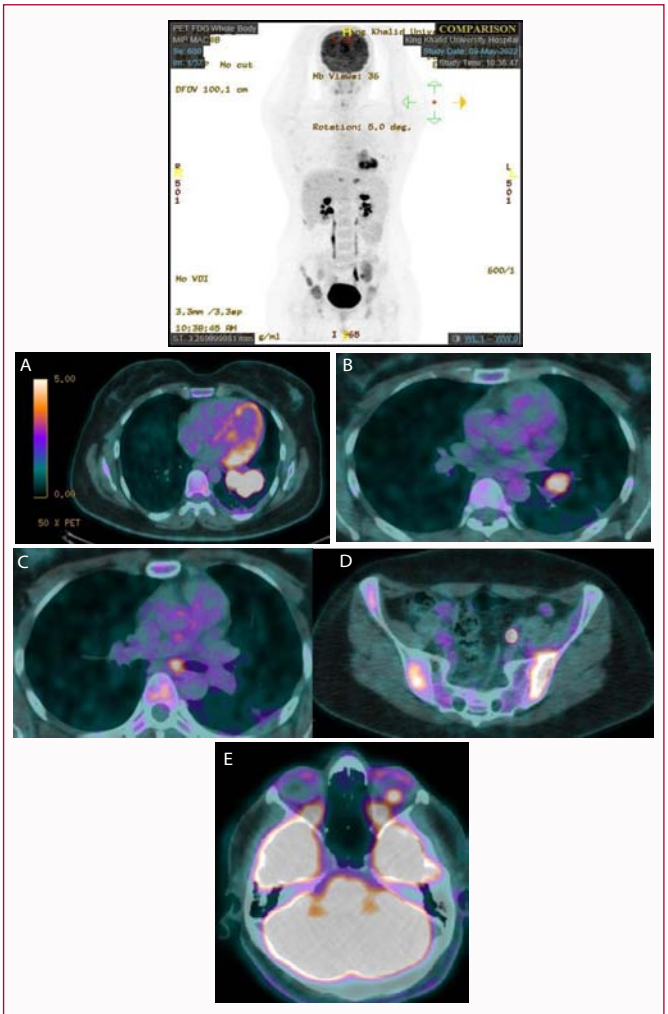

Figure 5: A) FDG-avid left lower lobe mass, SUVmax 16.4, with extension to the left hilum inferiorly, SUVmax 8.8. B) Mildly FDG-avid rounded atelectasis in left lower lobe. C) Small focus of FDG uptake in the left subcarinal region, likely lymph node, SUVmax 5.5. D) Multiple foci of uptake in the bone marrow with no corresponding osseous lesions. E) Small focus of FDG uptake in the posterolateral aspect of the left ocular globe, corresponding to the lesion seen on MRI, SUVmax 7.6.

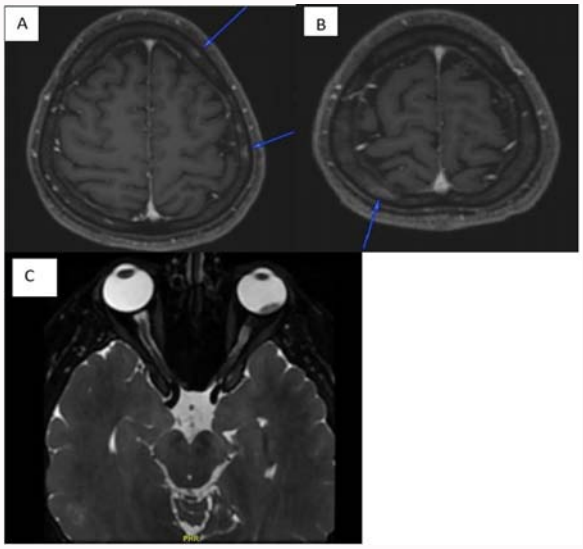

Figure 6: A&B) Multiple bilateral parietal and left frontal calvarial enhancing lesions likely metastasis. C) Left ocular enhancing lesion corresponding to the previously seen in PET-CT (Ocular metastasis).

showed progressive disease (Figure 6), CT chest and abdomen showed multiple hepatic and osseous metastases. Her chemotherapy regimen has been modified to carboplatin and Pemetrexed, Pembrolizumab remained unavailable. She is under active follow up with the medical oncology clinic.

Discussion

Adenoid cystic carcinomas has been noted to be histologically similar regardless of their primary sites. Three main growth patterns including cribriform, tubular, and solid are identified. Our patient had the cribriform subtype predominant pattern, which is the most frequent pattern observed in the literature [4]. Primary lung adenoid cystic carcinoma frequently occurs in the large airways, peripheral occurrence is very rare in the literature [5]. Our patient had involvement of the left main bronchus evident in the CT scan. These tumors usually affect patient between age 40 to 60, our patient is younger than the median age reported in case series [1].

Majority of reported cases present with respiratory complaints, the most frequent symptoms include cough, hemoptysis, or shortness of breath [1], none of which were present in our patient. Our patient presented to the ophthalmology clinic initially with complaint of reduction in vision due to choroidal metastasis, a presentation that has not been reported before in the literature for patients with adenoid cystic carcinoma of the lung yet to be diagnosed. Retinal metastasis has been reported in cases of primary adenocarcinoma and squamous cell carcinoma of the lung [6].

Our patient had stage IV disease, that progressed despite 4 cycles of chemotherapy, a behavior similar to reported cases in the literature [1]. These tumors are generally not sensitive to chemotherapy but may show partial response to targeted novel therapies, yet, more studies are required to establish the rule of targeted therapies. They may respond to radiotherapy as an initial therapy for resected or unresected advance stage [7]. In our patient, radiotherapy was used to palliate her back and hip pain from bone metastasis.

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